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Case Series

Microbiology Section

Mycetoma in Atypical Sites: A Case Series from Eastern India

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ABSTRACT

Mycetoma, a slowly progressive, chronic granulomatous infectious disease of the skin and subcutaneous tissues, is highly endemic in India. The triad of tumefaction, multiple draining sinuses, and grains in discharge clearly defines the disease. Mycetoma has now been declared a neglected tropical disease by the World Health Organisation (WHO). The disease is notoriously difficult to treat, characterised by stigmatising deformities, disabilities, and lifelong high morbidity. A strong clinical suspicion, appropriate diagnosis, and exact categorisation of the lesion up to the eumycotic or actinomycotic level, along with culture correlation, are essential for effective therapy of mycetoma. In this case series, authors report three cases of mycetoma manifesting in atypical sites of involvement. Two cases showed involvement of extremely unusual areas of the body, such as the shoulder and flank, while the third had lesions on the plantar aspect of the foot. One case was diagnosed as actinomycetoma caused by *Nocardia* spp., and two were diagnosed as cases of eumycetoma caused by *Madurella* spp. and *Exophiala* spp., respectively.

Keywords: Actinomycetoma, Eumycetoma, *Nocardia*

INTRODUCTION

Mycetoma is a slowly progressive, chronic granulomatous infection of the skin and subcutaneous tissues, often involving the underlying fascia and bones, and usually affecting the extremities [1].

The disease is characterised by a triad of:

1. Tumefaction of the affected tissue,
2. Formation of multiple draining sinuses, and
3. Presence of granules in the discharge.

These granules are tightly knit clusters of organisms within the infected tissue, also referred to as “grains” [1].

Mycetoma is notoriously challenging to treat. The most common sites for mycetoma are dorsum of the foot and the dorsum of the hands. Present series describes three cases of mycetoma at atypical sites like shoulder, flank and plantar aspect of foot respectively.

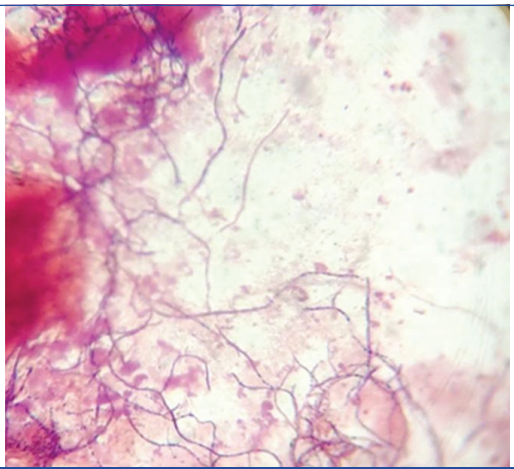
CASE SERIES

Case 1

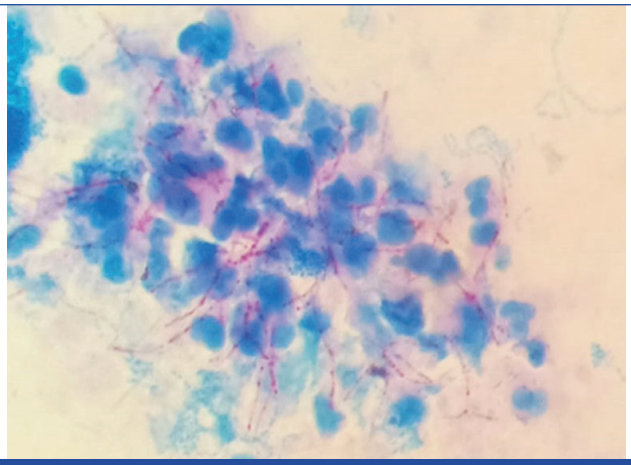
A 45-year-old lady from rural Bengal, a homemaker, presented to the surgical Outpatient Department (OPD) with multiple discharging painless ulcers over her left shoulder [Table/Fig-1]. She first noticed two nodular swellings four years ago, which have now progressed. There was discharge of serous fluid mixed with yellowish-white granules from the ulcers, which were soft to the touch. A few of these ulcers showed signs of healing. The patient could not recall any incidents of trauma or thorn-prick injuries to her left shoulder. However, she mentioned that she travels a long distance through a forest to fetch water and frequently carries the urn on her left shoulder. The patient was a known case of type II diabetes mellitus and has no history of hypertension.

Granules from the discharge were collected on a sterile petri dish by pressing the ulcers from the periphery with a sterile gauze piece. These granules were washed with sterile saline several times, crushed between slides, and inoculated on Sabouraud’s Dextrose Agar (SDA) and Lowenstein-Jensen (LJ) media. An X-ray of the left shoulder revealed no bony erosion. After 10 days, wrinkled, irregular, whitish-pink colonies appeared on the LJ media. Gram staining from the colony showed long filamentous gram-positive bacilli with branching [Table/Fig-2]. Ziehl-Neelsen staining with 1% H_2SO_4 as the decoloriser revealed long filamentous acid-fast bacilli [Table/Fig-3]. The pathogen was diagnosed as *Nocardia* spp., causing this case of actinomycetoma. Further speciation and antimicrobial susceptibility testing were not performed due to lack of facilities in our laboratory. The patient was treated with Amoxicillin and Cotrimoxazole, and the serous discharge reduced after eight weeks of treatment. The patient is still under follow-up.

[Table/Fig-1]: Multiple discharging painless ulcers over left shoulder.



[Table/Fig-2]: Gram stain showing long, filamentous gram positive bacilli with branching, under 100x magnification.



[Table/Fig-3]: Ziehl-Neelsen stain with 1% H_2SO_4 as decolouriser, showing long filamentous acid fast bacilli, under 100x magnification.

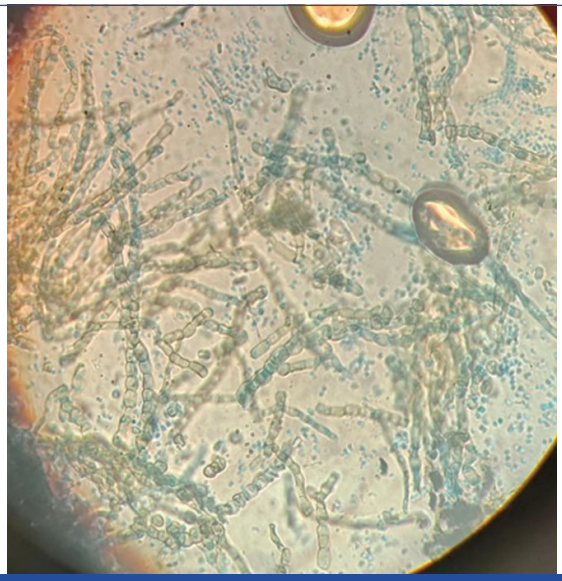
Case 2

A 19-year-old male college student from Kolkata came to the Dermatology OPD complaining of 5-6 very small, non healing, painless ulcers with occasional serous discharge on his left flank for one year [Table/Fig-4]. The lesions had initially started as small nodular swellings but progressively evolved into ulcers. There was no history of trauma; however, the teenager mentioned that he was a football player and spent long hours practicing or resting on the fields.

[Table/Fig-4]: Multiple healed ulcers on the flank of the patient.

The clinician referred the patient to the Microbiology department for investigation, suspecting actinomycosis or atypical mycobacterial infection. On gross examination, significant darkening of the overlying skin was noted. The discharge was subjected to Gram staining and culture-sensitivity testing, as well as ZN staining and KOH mount. Bacteriological culture showed no growth, and no microorganisms were found in the Gram stain, ZN stain, or KOH mount. After four weeks, a repeat sample was taken, during which a black grain was found along with pus. On direct examination with 10% KOH, round, firm to hard, black grains, approximately 0.5-1 mm in size, along with broad brown hyphae, were demonstrated. The crushed grain was then inoculated on SDA agar at 25 °C for two weeks. A greenish-yellow colony with radial grooves appeared on the SDA agar [Table/Fig-5], revealing brownish septate hyphae with intercalary chlamydospores on the Lactophenol Cotton Blue mount [Table/Fig-6]. The aetiological agent was diagnosed as *Madurella* spp. Identification was based purely on the morphological features of the colony and microscopic findings. The patient was given oral Itraconazole 400 mg per day, along with proper counseling about the prognosis of the disease. After six months, there was a slight reduction in skin darkening in the affected area and the amount of discharge.

[Table/Fig-5]: Greenish-yellow colonies of *Madurella* spp. on SDA.



[Table/Fig-6]: LPCB mount from *Madurella* spp. colonies showing brownish septate hyphae with intercalary chlamydospores, under 40x magnification.

Case 3

A 62-year-old male from a remote village in Nadia, West Bengal, a fisherman by occupation, presented to the Dermatology OPD complaining of a gradually increasing swelling on the plantar aspect of his left foot. He first noticed the swelling one year ago. The swelling had multiple discharging sinuses, and while it was initially painless, it became painful within 3-4 weeks. The patient had been advised to take antibiotics and analgesics by a local doctor, which led to healing of the sinuses, leaving puckered scars on his foot. However, after two months, the swelling reappeared, this time with even more sinuses discharging serous fluid and black grains [Table/Fig-7]. An X-ray of the foot revealed osteolytic changes [Table/Fig-8]. The

[Table/Fig-7]: Swelling on plantar aspect of left foot discharging serous fluid with black grains.

[Table/Fig-8]: X-ray of left foot of patient showing osteolytic lesions.

patient recalled accidentally stepping on a fishing hook and suffering a puncture wound under his left foot about three years ago.

Granules from the discharge were collected on a sterile petri dish. Direct KOH microscopy revealed soft, white grains around 300-400 µm in size, surrounded by plenty of thick fungal hyphae. After 10 days of incubation on SDA agar, a growth was diagnosed as *Exophiala* spp [Table/Fig-9]. The patient was prescribed oral Itraconazole at a dosage of 400 mg per day and showed significant improvement upon follow-up.

DISCUSSION

Mycetoma is commonly known as Madura Foot or Maduramycosis. There are broadly two categories of this entity: Eumycetoma, caused by fungi, and Actinomycetoma, caused by bacteria of the class Actinomycetes. It usually occurs through the introduction of the causative agent from saprophytic soil sources into the subcutaneous tissue via accidental trauma. The disease is endemic in areas with abundant vegetation and primarily affects field workers and farmers [1]. The WHO has now declared mycetoma as a neglected tropical disease [2]. Eumycetoma may be unresponsive to standard antifungal therapies, while actinomycetoma typically responds to antibiotic therapy, albeit with prolonged treatment required [3]. Despite receiving medical and surgical care, 25-50%

[Table/Fig-9]: *Exophiala* spp. colonies on SDA showing velvety grey appearance.

of individuals experience recurrences [4]. The exact onset of the disease may remain indeterminate, as patients often do not present until noticeable discomfort, swelling, or discharge occurs [5].

India is a high-endemic area for both types of mycetoma, with various manifestations presenting in this setting [6]. The most common sites for mycetoma remain the dorsum of the foot and the dorsum of the hands. However, in present study, mycetoma was found at unusual sites, such as the shoulder and flank, demonstrating that it can affect any part of the human body depending on the individual's habitat, working environment, and hygiene. Any penetrating injury from an agent contaminated with soil can cause the disease, regardless of immune status, age, or sex [7].

Whitish grains are typically produced by agents of actinomycetoma, while eumycetoma may produce both black and white grains depending on the causative agents [8,9]. Strong clinical suspicion, proper categorisation of lesions into eumycotic or actinomycotic, along with culture correlation, are essential for accurate prognoses and effective therapy [10]. In this case series, three different agents of mycetoma was isolated from atypical sites of involvement: one diagnosed as actinomycetoma caused by *Nocardia* spp and two diagnosed as cases of eumycetoma caused by *Madurella* spp (producing black grains) and *Exophiala* spp (producing white grains), respectively. The following table [Table/Fig-10] presents data on different cases of mycetoma studied over the past decade [6,11,12].

Author	Site of involvement in the body	Etiological agent isolated
Rit K et al., [11], 2015	Left foot	*Scedosporium apiospermum*
Nupur P et al., [6], 2020	Left foot and Left buttock in the same patient	*Acremonium falciforme* and *Madurella grisea*
Cazzato G et al., [12], 2021	Right foot	*Streptomyces somaliensis*
Mandal S et al., 2026	Left shoulder Left flank Left foot	*Nocardia* spp. *Madurella* spp. *Exophiala* spp.

[Table/Fig-10]: Showing data on different cases of mycetoma (arranged year-wise) [6,11,12].

CONCLUSION(S)

Mycetoma is a debilitating disease characterised by profound deformities and disabilities that significantly affect the patient's quality of life. If left untreated, it gradually progresses to involve the underlying bone, muscle, and other adjacent tissues. The common presentation sites for mycetoma in the human body are the dorsum of the foot and hands—areas frequently exposed to penetrating injuries (such as thorn pricks or wounds). However, present study found mycetoma at unusual sites, demonstrating that it can affect any part of the human body, depending on individual habitats, everyday routines, occupations, or accidental penetrating injuries by agents contaminated with soil. Since the disease typically presents months or years after the relevant trauma, the patient's recollection may not always be reliable. Therefore, in the evaluation of a swelling with multiple discharging sinuses at any body part, the possibility of mycetoma should always be considered.

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